TABLE 1. Associations of Castleman Disease With Immunologically Mediated and Other Systemic Diseases

Systemic Conditions
Myasthenia gravis
Severe erosive lichen planus/stomatitis
Pemphigus vulgaris
Temporal arteritis
Monoclonal hypergammaglobulinaemia, hypoalbuminaemia and refractory anemia (plasma cell variant)
Thrombocytopenic purpura (hyaline vascular variant)
Nephrotic syndrome
POEMS syndrome (polyneuropathy, organomegaly, endocrinopathy, M proteins and skin changes)
HIV
Kaposi sarcoma
Lymphoma
Vascular neoplasms

POEMS, polyneuropathy organomegaly endocrinopathy and M protein and skin changes.

confirms the importance of a careful clinical examination (by the oral surgeon/dentist) in providing a good healthcare. The primary lesion was resembling other more common oral lesions like pleomorphic adenoma, lymphoma, and nodular necrotizing sialometaplasia. The definitive diagnosis of the CD, in this patient report, was histopathological. Thus, the clinical examination and the histopathological analysis have been the key factors in detecting rare and uncommon pathologies in the oral cavity.

Castleman disease rarely occurs in the head and neck region.[8] It may appear in any lymph node but most commonly in the mediastinum.[6,7,9] Clinically, there are 2 forms of CD: unicentric (the most common) and the multicentric forms. The former is usually presented in a lymphonodular region while the latter is often associated with fever, diaphoresis, fatigue, weight loss, and even with some malignancies including Kaposi sarcoma, non-Hodgkin lymphoma, Hodgkin disease, and polyneuropathy organomegaly endocrinopathy and M protein and skin changes.[7–11]

Histologically, there are 2 types of solitary CD: the more common hyaline-vascular type and the plasma cell type. The hyaline-vascular type is not usually associated with systemic nor hematologic manifestations and it is the most common variant in the head and neck region (98%).[6–8] Whereas patients with the plasma cell type may present a wide variety of symptoms and disorders such as fever, anaemia, polyclonal hypergammaglobulinemia, nephritic syndrome, and splenomegaly. This aggressive variant occasionally progresses to lymphoma, conveying a poor prognosis. It has been described the association of CD with immunologically mediated and systemic diseases (Table 1). Some of these conditions contraindicate certain dental treatments, such as implants.

Surgical resection is the treatment of choice for cervical CD. It has been reported a 100% 5-year control rate for the HV type of unicentric CD.[9] In our patient, recurrence is usually uncommon if the excision of the mass is complete.

CONCLUSION

Careful clinical examination and histopathological analysis of a biopsy are essential for the diagnosis of rare and unusual pathological changes of the oral mucosa. A new patient of hyaline-vascular unicentric CD has been diagnosed in the hard palate.

ACKNOWLEDGMENT

The authors thank Dr. José Manuel Aguirre Urizar for the histopathological analysis.

REFERENCES

1. Neville BD, Damm DD, Allen CM, et al. Oral manifestations of systemic diseases. In: Neville BD, Damm DD, Allen CM, Bouquot JE, eds. *Oral and Maxillofacial Pathology.* St. Louis, MO: Saunders Elsevier; 2009:761–793
2. Castleman B, Iverson L, Mendez VP. Localised mediastinal lymph node hyperplasia resembling thymoma. *Cancer* 1956;9:822–830
3. Anagnostou D, Harrison CV. Angiofollicular lymph node hyperplasia (Castleman). *J Clin Pathol* 1972;25:306–311
4. Keller AR, Hochholzer L. Castleman B Hyaline vascular and plasma cell types of giant lymph node hyperplasia of the mediastinum and other locations. *Cancer* 1972;29:670–683
5. Dupin N, Diss TL, Kellam P, et al. HHV-8 is associated with a plasmablastic variant of Castleman disease that is linked to HHV-8-positive plasmablastic lymphoma. *Blood* 2000;95:1406–1412
6. Maruyama S, Hao N, Cheng J, et al. Castleman's disease of the buccal mucosa: report of a case and review of the literature of head and neck cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;93:305–310
7. Kilty SJ, Yammine NV, Corsten MJ, et al. Castleman's disease of the parotid gland. *J Otolaryngol* 2004;33:396–400
8. Gürbüzler L, Ceylan A, Yilmaz M, et al. Castleman's disease of the parotid gland: a case report. *Kaohsiung J Med Sci* 2010;26:444–447
9. Zhong LP, Chen GF, Zhao SF. Cervical Castleman disease in children. *Br J Oral Maxillofac Surg* 2004;42:69–71
10. Sanchez-Cuellar A, de Pedro M, Martin-Granizo R, et al. Castleman disease (giant lymph node hiperplasia) in the maxilofacial región: a report of 3 cases. *J Oral Maxillofac Surg* 2001;59:228–231
11. Samadi DS, Hockstein NG, Tom LW. Pediatric intraparotid Castlemans's disease. *Ann Otol Rhinol Laryngol* 2003;112:813–816

Removal of an Unusual Neglected Foreign Body in Infratemporal Region Using Navigation

Avani Jain, MS, Gaurav Gupta, MS,† and Mohnish Grover, MS‡*

Abstract: A 19-year-old male presented with complaint of a bluish mass in the hard palate since 3 months. The patient had a history of trauma 8 years back in the left zygomatic area with a pen. It was lodged in the wound and removed at that time. Computed tomography scan was revealed a linear heterogenous dense structure extending from left infratemporal fossa to oral cavity, traversing through left maxillary sinus, with bone defect seen in lateral and medial wall of maxilla, and in the hard palate, most likely a

From the *Department of Otolaryngology (ENT), MAMC & Lok Nayak Hospital, New Delhi; †Department of Otolaryngology (ENT), PBM Hospital, Bikaner; and ‡Department of Otolaryngology (ENT), SMS Hospital, Jaipur, Rajasthan, India.

Received September 6, 2016.
Accepted for publication October 21, 2016.

Address correspondence and reprint requests to Dr Avani Jain, MS, Flat No C-8, Tower 1, New Moti Bagh, New Delhi 110023, India; E-mail: avanijain87@hotmail.com

The authors report no conflicts of interest.

ISSN: 1049-2275
DOI: 10.1097/SCS.0000000000003402

neglected foreign body. The foreign body was removed by navigation-assisted endoscopic surgery and the palatal perforation repaired using local rotation flap. There were no intraoperative or postoperative complications. Navigation-guided removal of foreign body in proximity to vital structures, in the infratemporal region, is a valuable option with minimal morbidity.

Key Words: Foreign body, image-guided surgery, infratemporal fossa

Foreign body in the aerodigestive tract is a common emergency encountered in otolaryngology practice. However, the lodgment of a foreign body in the infratemporal fossa is rare and only a few patients have been reported in the literature. Its removal is a challenge due to critical structures associated with the infratemporal fossa. Here we report an interesting patient with a long-standing neglected foreign body in the infratemporal fossa, which migrated to the palate and was eventually removed using navigation-assisted endoscopic surgery. As per our knowledge, this is the first patient to be reported of such a long standing, migrating foreign body that was removed with the help of navigation system.

CLINICAL REPORT

A 19-year-old male presented to the otorhinolaryngology out patient department, with the complaint of a bluish mass seen in the center of the hard palate. On taking detailed history, the patient had a history of assault in the left zygomatic area with a pen 8 years back. A part of the pen that had got lodged in the wound was removed at a local hospital under local anesthesia at that time and the wound healed on its own. The patient was asymptomatic following the incident. However, since 3 months, the patient noticed a bluish mass in the palate. On examination, a foreign body could be seen perforating through the hard palate in the oral cavity (Fig. 1A). There was also a healed scar in the left zygomatic area, which most likely represented the entry wound of the pen. Diagnostic nasal endoscopy was found to be normal; no foreign body could be identified. A computed tomography (CT) scan was advised, which revealed a linear heterogenous dense structure measuring approximately 6 cm in length, extending from left infratemporal fossa to oral cavity, traversing through left maxillary sinus, with bone defect seen in lateral and medial wall of maxilla, and in the hard palate, most likely a neglected foreign body.

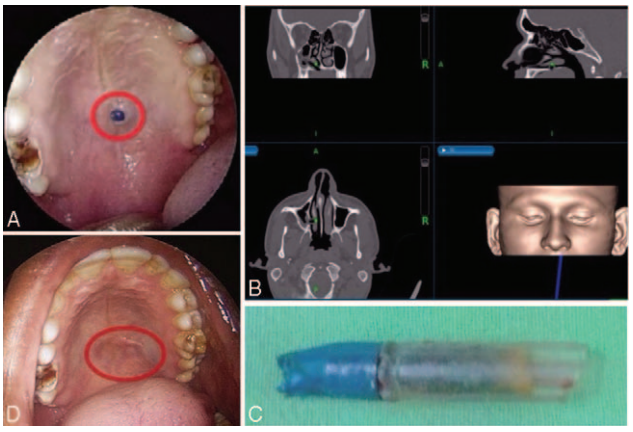

FIGURE 1. (A) Exit wound: Foreign body (pen) seen perforating through the hard palate, (B) navigation-assisted localization of foreign body, (C) foreign body part of a pen removed as a single piece, and (D) healed palatal wound 1 year after follow-up.

An attempt was made to remove the foreign body through the palate, but it was found to be fixed because of adhesions to adjacent structures. The patient was planned for exploration and foreign body removal under general anesthesia, by navigation-assisted endoscopic surgery. Written informed consent was taken prior to surgery. We used Medtronic S7 stealth station navigation and fusion system, which is based on advanced optical camera technology. Using the navigation probe, the foreign body was located (Fig. 1B) and an incision was made in the posterior third of left inferior turbinate anterior to the level of pterygoid. The foreign body was located and gently adhesions were released. With the navigation, full course of the foreign body was tracked and delivered as a single piece through the nose. It was a broken piece of pen measuring approximately 5 cm in length (Fig. 1C). After removal of the foreign body, the margins of the palatal perforation were freshened. The palatal perforation was repaired using local rotation flap. The patient was given intravenous antibiotic for 5 days following surgery and was discharged on oral antibiotics. No foreign body was noted at the follow-up facial CT scan at 1 month. After 1 year follow-up, the patient is asymptomatic with a well-healed palate wound (Fig. 1D).

DISCUSSION

In the past few decades, with the increase in the rates of assault and trauma such as road traffic accidents and gunshot wounds, there has been an increase in the penetrating foreign body injuries in the maxillofacial region. The infratemporal fossa is a compact space and many important nerves and vessels traverse it. As important organs like brain and eye are in the immediate vicinity of it, and this space communicates with these organs via anatomical foramina, there is a potential for the foreign body and associated inflammatory reaction to migrate and cause life threatening complications like hemorrhage, blindness, and various intracranial complications.[1] Complications arising from these foreign bodies usually arise early in the postinjury period due to their mass effect, and later because of infection. In most of the patients with foreign body in the infratemporal reported in the literature, the cause was trauma and the main symptom being trismus.[2,3] Park Do et al reported a rare patient with infratemporal cellulitis due to iatrogenic surgical gauze.[4] A unique feature of our patient was that it presented as a diagnostic dilemma as there was a false sense of security that the foreign body was removed at the initial time of presentation. The onset of clinical symptoms was 8 years after the initial penetrating injury. Apart from the delay in presentation, the other unusual feature in our patient was the migration of the foreign body from the infratemporal fossa to the hard palate. To date, an asymptomatic neglected foreign body in the infratemporal fossa migrating and perforating the hard palate has not been described in the literature.

It has been suggested that all foreign bodies that are symptomatic and those that are located near vital structures warrant removal.[5] The method of removal is controversial and various techniques have been described in literature. The external approaches to this area are classified as lateral (transzygomatic and lateral infratemproal), inferior (transmandibular and transcervical), or anterior (transfacial, transmaxillary, transoral, and transpalatal).[6] Removal of foreign body from infratemporal fossa is a surgical challenge as it is an inaccessible location and also there are important surrounding structures, such as the nerves and carotid artery. Identifying the location of the foreign body and determining a safe surgical approach are difficult using conventional radiography, particularly in patients with radiolucent foreign body like ours. Image-guided navigation technology has been used successfully in the management of various skull base tumors. However, there are very few reports on the use of navigation for foreign body

removal.[5,7] An image-guided navigation system allows for preoperative planning and intraoperative visualization of the foreign body and its relation with the surrounding structures. It provides the advantage of improved surgical accuracy, shortened operating time, and minimally invasive access thereby reducing the overall morbidity as well as duration of hospitalization.[5] In our patient, with the aid of the navigation system we were able to localize and remove a large foreign body (part of pen) through the nose, thereby avoiding injury to major structures and an external scar, and thereby reducing the overall morbidity.

REFERENCES

1. Grant CA, Rubin PAD. An infratemporal fossa foreign body presents as an infraorbital. *Mass Arch Ophthalmol* 2000;118:993–995
2. Purohit JP, Kumar G, Singh PN, et al. An unusual foreign body in infratemporal fossa. *Indian J Otolaryngol Head Neck Surg* 1996;48: 323–324
3. Thakur JS, Chauhan CGS, Diwana VK, et al. Trismus: an unusual presentation following road accident. *Indian J Plast Surg* 2007;40: 202–204
4. Park Do Y, Choo OS, Hong SY, et al. Infratemporal fossa cellulitis caused by a remnant iatrogenic foreign body after a bimaxillary operation. *J Craniofac Surg* 2015;26:e196–e197
5. Gui H, Yang H, Shen SG, et al. Image-guided surgical navigation for removal of foreign bodies in the deep maxillofacial region. *J Oral Maxillofac Surg* 2013;71:1563–1571
6. Neff LL, Liess BD, Chang CW. Transoral endoscopic removal of a bullet from the infratemporal fossa. *Otolaryngol Head Neck Surg* 2008;138:113–114
7. Koo Ng NK, Jaberoo MC, Pulido M, et al. Image guidance removal of a foreign body in the orbital apex. *Orbit* 2009;28:404–407

Transcutaneous Ultrasonography for Diagnosis of Nasolabial Cyst

Chung-Hui Yeh, MD, Jenq-Yuh Ko, MD, PhD,† and Cheng-Ping Wang, MD†*

Abstract: Nasolabial cyst is usually diagnosed by computerized tomography and magnetic resonance imaging. Ultrasonography could be a simple, office-based diagnostic imaging tool for nasolabial cyst. The authors report a 46-year-old woman with painful swelling on the bilateral anterior nostrils. The high-frequency (13 MHz) linear array transducer of the ultrasound (Hitachi Hivision Avius with EUP-L74M, Hitachi Aloka Medical, Tokyo, Japan) was transversely placed on the upper lip to scan the anterior nasal floor. Ultrasonography showed 2 well-defined anechoic oval cystic lesions in the anterior nasal floor, which were compatible with nasolabial cysts. Magnetic resonance imaging, which was done for her nasopharyngeal carcinoma 3 years ago, showed the same cystic lesions in the anterior nostril floor. Transcutaneous ultrasonography could be a simple, first-line imaging tool for diagnosis of nasolabial cyst in an office-based clinical setting.

Key Words: Cyst, nasal cavity, nasolabial, nose, ultrasound

Nasolabial cyst is an uncommon nonodontogenic cystic lesion on apices of incisor of maxilla. The most common clinical features are facial swelling (70.9%) and nasal obstruction (17.3%).[1–4] The symptomatic cysts are typically present in the fourth and fifth decades. Imaging study is usually necessary for diagnosis including computerized tomography (CT) and magnetic resonance imaging (MRI) that are the most common tools to confirm the diagnosis. However, it is not convenient for CT or MRI to diagnose this relatively simple disease in the clinical practice. Because nasolabial cyst is located in the most anterior nasal floor, without bony structure blocking the ultrasound energy transmitting from the anterior upper lip skin to the lesion, ultrasonography (US) may be an alternative simple, first-line imaging tool to detect nasolabial cyst in an office-based clinical setting.

Here, we presented a patient with nasopharyngeal carcinoma, who had infected bilateral nasolabial cysts, which were diagnosed by transcutaneous US and confirmed by reviewing previous MRI for staging nasopharyngeal carcinoma 3 years ago.

CLINICAL REPORT

A 46-year-old woman with nasopharyngeal carcinoma, who completely treated 3 years ago, presented with painful swelling over the anterior nostrils for several days. She found upper lip asymmetry after the pus was coming out from the right nasal cavity (Fig. 1A). On physical examination, the left nasal floor was elevated with partial obstruction of the left anterior nostril, compared with the right nose. Some pus-like substance could be squeezed out from the small ruptured wound on the right anterior nasal floor. Ultrasonography examination with a high-frequency (13 MHz) linear array transducer (Hitachi Hivision Avius with EUP-L74M, Hitachi Aloka Medical, Tokyo, Japan) placed transversely on the upper lip close to the anterior nostril showed 2 well-defined, regular-shaped, homogenous, anechoic, avascular oval cystic lesions abutting the maxilla bone with little solid component and with a strong acoustic enhancement line between the posterior cystic wall and the bone. The swollen skin tissue above the cystic lesions was also noted on US (Fig. 1B). The right anechoic cyst was much smaller than the left one because of rupture of the cyst.

Magnetic resonance imaging, which was done for nasopharyngeal carcinoma 3 years ago, revealed bilateral well-defined cystic lesions in the anterior nasal floor, with homogeneous hypointensity on T1-weighted image and hyperintensity on T2-weighted image (Fig. 1C). Excision of bilateral nasolabial cysts was suggested, but she decided to receive regular observation instead of surgical intervention.

DISCUSSION

Nasolabial cyst, also called nasoalveolar cyst or Klestadt cyst, is a rare nonodontogenic cystic lesion on the apices of incisor of maxilla.[5,6] Most nasolabial cysts are unilateral, especially left side. Only 10.9% are bilateral.[1] The mean age at diagnosis is in the fourth and fifth decades, with female-to-male ratios of 4:1–3:1.[1,7–9]

The diagnosis of nasolabial cyst is based on clinical features, imaging studies, and histology. The most common image studies

From the *Department of Otolaryngology, Taichung Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, Taichung; and †Department of Otolaryngology, National Taiwan University Hospital and National Taiwan University, College of Medicine, Taipei, Taiwan.

Received September 22, 2016.
Accepted for publication October 21, 2016.

Address correspondence and reprint requests to Cheng-Ping Wang, MD, Department of Otolaryngology, National Taiwan University Hospital, No. 7, Chung-Shan South Road, Taipei 100, Taiwan;
E-mail: wangcp@ntu.edu.tw

The authors report no conflicts of interest.

ISSN: 1049-2275
DOI: 10.1097/SCS.0000000000003403